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Case Report

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Fibrous Dysplasia in Paranasal Cavities

I. Three Case Reports

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Key Words
Fibrous dysplasia
Sinus

Abstract
Presentation is made of 2 boys and a girl with fibrous dysplasia involving the frontal and ethmoidal sinuses and the maxilla. This tumor-like growth of the bone was not restricted to the sinus alone but expanded to the orbit, adjacent sinus, skull base and infratemporal or pterygoid fossa. In such cases, surgical therapy is the treatment of choice, with maximal preservation of healthy tissue and avoidance of major devastations that may result in undesired cosmetic defects. The etiology of the disease is still unknown. However, it should be emphasized that all 3 patients had suffered an injury to the adjacent bony structures at various time points preceding the disease manifestation. That is why we are inclined to believe that trauma might be a factor responsible for the onset of fibrous dysplasia.

Introduction

Fibrous dysplasia is a benign lesion of the bone, characterized by substitution of the normal bony structure by fibrous-osseous connective tissue, which histologically represents various stages of bone metaplasia [1]. Proportions of the fibrous and osseous tissues vary, occasionally even in the zones of the same bone [2–4]. Furthermore, recent data have pointed to a relationship between the preexisting primary bone lesion, such as fibrous dysplasia, and an aneurysmal bone cyst of the paranasal sinuses [5] or a mucocele [6].

Various classifications of the disease have been proposed. The one suggested by Ramsey et al. [7] appears to be most convenient, describing three types of the disease: (a) monostotic, single or multiple lesions in the same bone, (b) polystotic, multiple lesions affecting more than one bone and (c) disseminated, multiple lesions, occasionally accompanied by extraskeletal involvements.

Considering the craniofacial complex as a whole, the mandible and maxilla are the most frequently described localizations of the disease [1, 8–12].

The etiology of the genesis and development of fibrous dysplasia has not yet been clarified.

Material and Methods

Patients

Case 1. A boy, A.I, aged 12. A year before admission to the hospital, a gradual prolapse of the left eye from the orbit, anterolaterally, accompanied by gradual impairment of the left eye vision, was observed (fig. 1): visus sin. 0.31–0.50, dsph. 0.9, visus dex. 1.0. Other examinations: X-ray tomography and CT showed a solid bone-like shadow along the anterior and posterior ethmoids, radiating into the left frontal sinus and left orbit, and pushing the bulbus anterolaterally (fig. 2). Perioperative findings revealed the tumor to have involved the mentioned sinuses, spreading into the orbit and pressing against the bulbus and optic nerve. The diagnosis of fibrous dysplasia was histologically confirmed. A drain through the nose was left for 3

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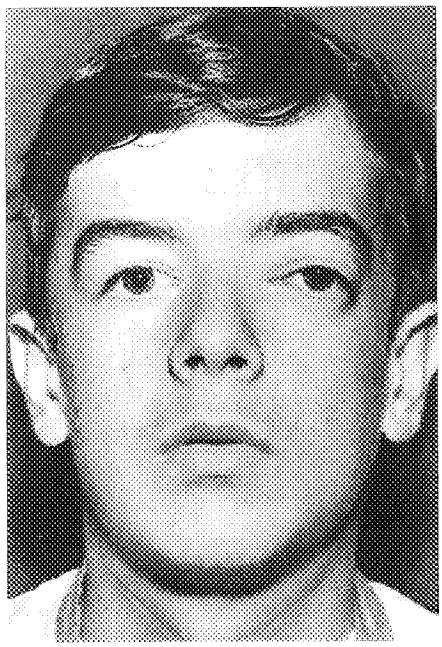

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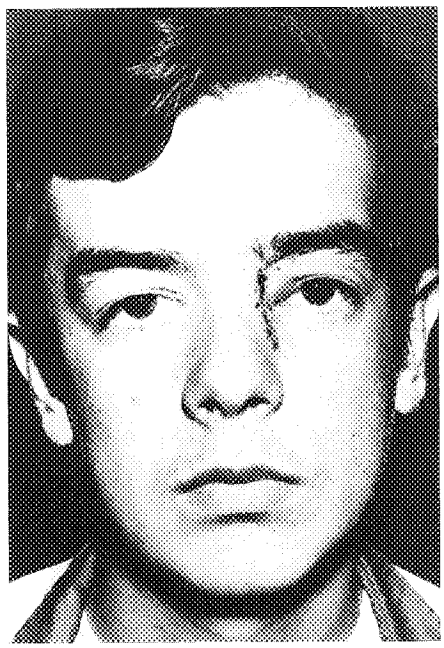

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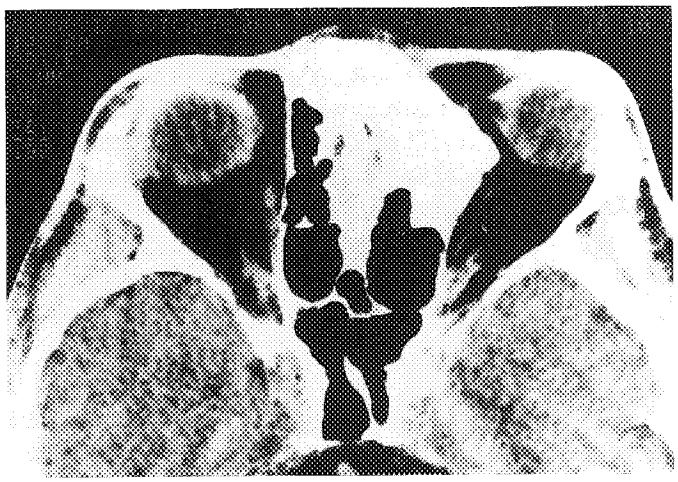

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Fig. 1. A boy, A.I., with the lesion localized in the left ethmoidal and frontal sinuses, spreading into the orbit. Discrete anterolateral dislocation of the left bulb.

Fig. 2. CT scan displaying the lesion localized in the left ethmoidal sinus, spreading into the frontal sinus, nose and orbit, with dislocation of the bulb and the lesion pressing against the optic nerve.

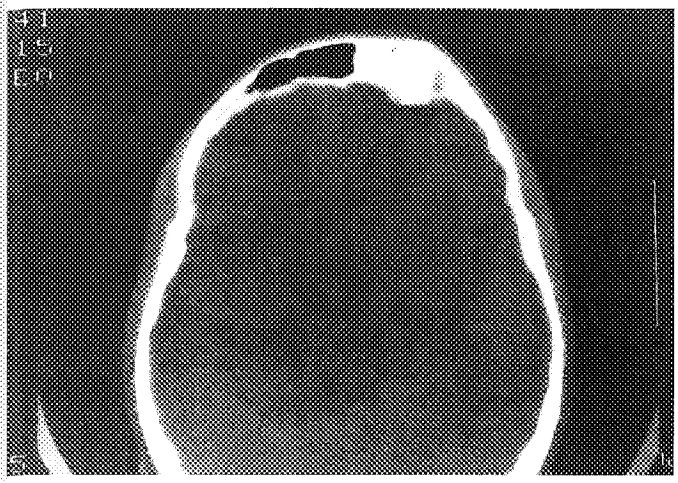
Fig. 3. The same patient, 3 weeks after surgery. The left bulb has resumed its normal position.

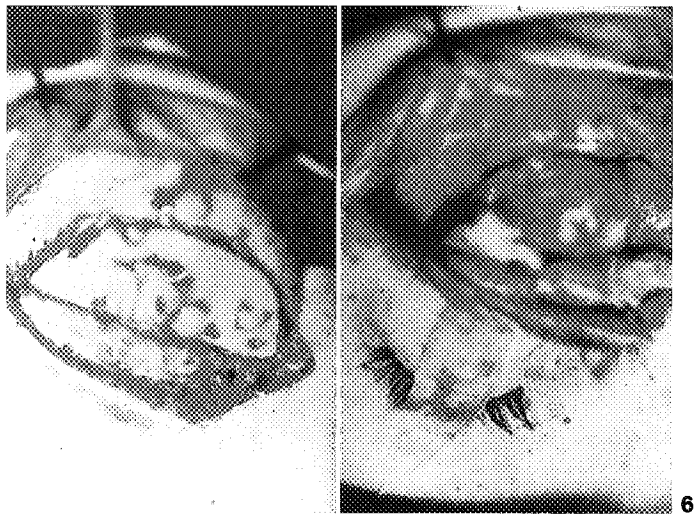
Fig. 4. Native X-ray taken in the other boy, V.E., shows fibrous dysplasia to be localized in the right frontal sinus, with minor involvement of the ethmoidal sinus.

weeks. Two weeks after the surgery, vision control showed the left eye sight considerably improved: 0.8–0.75, dsph. 1.0 (fig. 3).

Case 2. A boy, V.E., aged 15. A year before admission to the hospital, a hard swelling was observed in the right supraorbital region, which grew gradually and very slowly. Native X-ray and CT revealed a bone-like shadow occupying almost the entire right frontal sinus and spreading into the anterior right ethmoidal sinus (fig. 4, 5). The tumor was operatively found and completely removed, and extensive nasal drainage of the thus freed cavity was left for 3 weeks (fig. 6). The anterior lamina of the frontal sinus, partially perforated by the tumor process, was used for reconstruction of the anterior sinus wall, placing it over the sutured periosteum. Both the cosmetic and functional effects were very good.

Case 3. A girl, R.A., aged 11. A slowly growing tumor was observed 7–8 months before admission to the hospital. CT examination revealed a large bone-like tumor completely filling the right maxillary sinus (fig. 7). The tumor was removed by a procedure according to Caldwell-Luc. The operative field was allowed to drain into the nose.





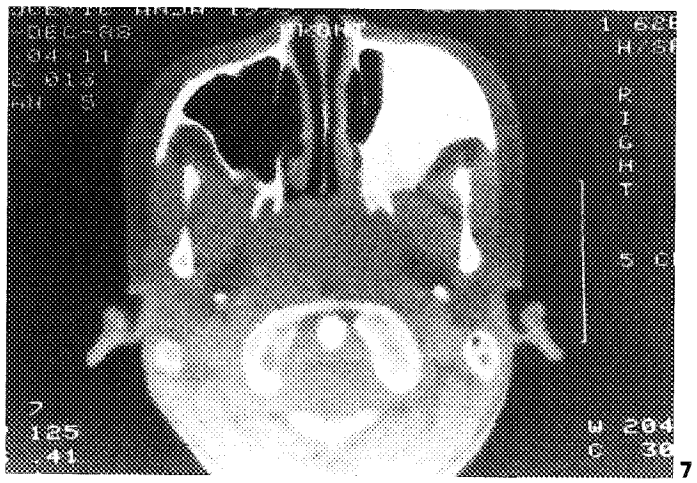


Fig. 5. Frontal sinus CT in the same boy, showing projection of the lesion into the right sinus, with marked deepening of the sinus due to tumor growth.

Fig. 6. The same patient. Section through the anterior sinus lamina and bone perforated by tumor process. The left lamina was removed. The tumor is visible.

Fig. 7. A girl, R.A., with fibrous dysplasia localized in the right maxillary sinus. CT scan.

Discussion

Three cases of fibrous dysplasia into the ethmoid, frontal sinus and maxilla are described. All three patients were young (11, 12 and 15 years of age). Ramsey et al. [7] have reported on 47 patients (age range, 3–37 years) with the disease. The majority of their patients (83%) were aged ≤ 20 years.

In our study, all 3 patients had previously suffered a trauma to the region adjacent to the site of dysplasia. The first boy was kicked into the left temporal region while playing football 2 years before the disease manifestation. The other boy was hit in a fight by a stick into the right frontoparietal region, whereas the girl fell during a gym class, with a bump against her right temporozygomatic region. During the posttraumatic period, late manifestations in the form of tumor-like alterations of large dimensions occurred, making the surgical procedure quite difficult.

In the management of these lesions, the surgical procedure may also entail a cosmetic defect. In each of the three cases reported, both the functional and cosmetic effects of the operation were quite satisfactory. In the first patient, the eye resumed its normal position in the orbit, and the vision improved considerably. In general, isolated visual dyfunction, with or without proptosis, has rarely been described as a sign of benign sphenoethmoid lesions [12]. In the second case, reconstruction of the anterior wall of the frontal sinus by the anterior lamina produced a very good result, whereas in the third case the right cheek remained slightly deformed, due to the removal of the anterior wall of the maxilla.

The etiology of the disease is unknown, with endocrine abnomalities, development from a hamartoma, trauma

and defects in bone developmentsuggested as the possible causes [3, 13, 14].

Presentation of our cases and the history of trauma as a possible initiating factor for the occurrence of the pathology described may hopefully contribute to the understanding of the etiology of this disease.

As differentiated from most bone and odontogenic tumors, fibrous dysplasia requires extensive local removal, including partial maxillectomy to obtain clear margins [7]. The surgical procedure should be thus designed to allow the development of dentition and facial developmental centers in the palate, nose and zygomatic ending of the maxilla [15].

Due care should be taken when deciding on the radicality of the procedure, because it is very difficult to determine the real border between the normal bone and the newly formed bone-like tumorous growth. Great caution and use of an operating microscope in all questionable steps of the operation are therefore recommended.

Following removal of major lesions, drainage of the newly formed spaces and cavities should be allowed for a prolonged period of time, i.e. for at least 3 weeks. We consider this period to be long enough to allow new epithelization and formation of a good drainage tube.

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